

CASE REPORT

Aortoesophageal Fistula with a History of Graft Treatment for Thoracic Aortic Aneurysm

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Aortoesophageal fistula (AEF) is a rare but fatal cause of gastrointestinal hemorrhage, being most commonly associated with thoracic aortic aneurysm. We present a rare case of secondary AEF post graft treatment for mycotic aneurysm of thoracic aorta 4 years previously. Massive upper gastrointestinal hemorrhage resulted in difficult diagnosis by gastrointestinal endoscopy. Computed tomography angiography (CTA) showed contrast extravasation from the descending aorta into the mid-third esophagus. CTA is a useful diagnostic tool in patients suspected of having AEF. Rapid identification of AEF is important for definitive treatment and increases the likelihood of survival. [*J Chin Med Assoc* 2008;71(2):100–102]

Key Words: aortic aneurysm, fistula, gastrointestinal hemorrhage

Introduction

Aortoesophageal fistula (AEF) is a rare but fatal cause of massive upper gastrointestinal hemorrhage. Thoracic aortic aneurysm is the most common cause of primary AEF,¹ while other causes include foreign body ingestion, esophageal carcinoma, trauma and tuberculous aortitis.^{2–4} Secondary AEF is rarely reported after endoscopic esophageal procedures, operative procedures and endoluminal stenting.^{1,5} AEF is usually identified postmortem after an exsanguinating hemorrhage.⁶ Early diagnosis and emergent surgical intervention are mandatory for survival. Here, we report a patient who presented with persistent hypotension and intermittent hematemesis, who had AEF with graft replacement for a mycotic thoracic aorta aneurysm over a 4-year period.

Case Report

A 78-year-old male was brought to the emergency department because of 1 episode of hematemesis in the

morning. He presented with acute onset of chest pain, nausea, and an episode of small-volume hematemesis. He had a history of mycotic aneurysm of the descending thoracic aorta post segmental replacement with a stent graft (Ultramax: 22 mm in diameter, 10 cm in length). Tissue culture obtained from surgical debris yielded *Salmonella*, group D.

Initial vital signs showed a temperature of 36.2°C, heart rate of 108 beats/minute, blood pressure of 72/42 mmHg, and a respiratory rate of 18 breaths/minute. Physical examination revealed an acute, ill-looking elderly man, who appeared uncomfortable and slightly disoriented, with an operation scar over his left upper back area. Because of persistent hypotension and intermittent hematemesis, initial resuscitation including endotracheal intubation for airway protection was started. Blood transfusion with 4 units of packed red blood cells and 2 units of whole blood was given within 8 hours. Chest X-ray revealed tortuosity of the thoracic aorta with cardiomegaly and widening of the mediastinum. Laboratory findings included white blood cell count of 42,900/mL, with 93.5% neutrophils, 1.4% lymphocytes and 5.1% monocytes, hemoglobin



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level of 9.1 g/dL, platelet count of 211,000/mL, blood glucose of 177 mg/dL, blood urea nitrogen of 33 mg/dL, creatinine of 2.4 mg/dL, sodium of 144 mmol/L, and potassium of 3.6 mmol/L.

Upper gastrointestinal endoscopy was performed with a standard forward-viewing endoscope with a 2.8-mm diameter accessory working channel. It revealed much fresh blood and hematoma within the esophagus, stomach, and duodenum, but the source of the bleeding could not be identified. Subsequent computed tomography angiography (CTA) demonstrated contrast extravasation from the descending aorta into the mid-third esophagus (Figure 1). The contrasted blood flowed down into the esophagogastric junction and fundus of the stomach (Figure 2). CTA also showed abnormal adherence of the esophagus to the

aorta and the presence of gas around the aortic prosthesis (Figure 1). Emergency surgical intervention was planned. However, cardiorespiratory resuscitation and blood transfusion were unsuccessful and his vital signs deteriorated progressively. The patient was then discharged against medical advice due to refractory hypovolemic shock.

Discussion

AEF is a relatively rare but life-threatening cause of upper gastrointestinal bleeding, constituting less than 10% of all aortoenteric fistulas.¹ Thoracic aortic aneurysms are the most common pathogenic cause of AEFs, and other pathogenic conditions, including atherosclerosis, infectious disease, carcinoma, Barrett's ulcer, foreign bodies, and complications of prolonged nasogastric tube intubation, are also reported to be associated with primary fistulas.¹ Secondary AEFs are defined as those that follow a previous operation and are a well-recognized complication of aorta prostheses. The pathogenesis of secondary AEF is still controversial, but both mechanical and infective factors may play an important role in their development. Our case had a history of mycotic thoracic aneurysm post graft replacement 4 years previously. Complete blood count showed severe leukocytosis with left shift. Two sets of blood cultures obtained in the emergency department yielded *Citrobacter freundii* and *Streptococcus sanguinis* 4 days later. Therefore, an association with graft infection cannot be ruled out. The presentation of both primary and secondary AEFs can be highly variable. Presentation with the classic Chiari's triad (dysphagia or mid-thoracic pain and initial sentinel hemorrhage, followed by exsanguinations) is usually diagnostic.⁷ In a review of 500 cases of AEF, 59% had mid-thoracic pain, 45% experienced dysphagia, 65% had herald bleeding, and 45% showed Chiari's triad.¹ The need for a high index of suspicion coupled with rapid evaluation is underscored by the fact that most cases of AEF are diagnosed postmortem.

Timely diagnosis of AEF usually involves 1 or a combination of imaging studies. In a series of 78 cases of primary AEFs, the chest X-rays were reported to be abnormal in only 32%.⁸ Barium swallow examination might show the thoracic aortic aneurysm as an extrinsic compression and deviation.⁹ Aortography is used for diagnosing the underlying aortic pathology, although the fistula itself is invariably not visible.⁹ Aortography also provides the opportunity for endovascular aortic stenting in patients who are not actively bleeding. Upper gastrointestinal endoscopy is recognized as the

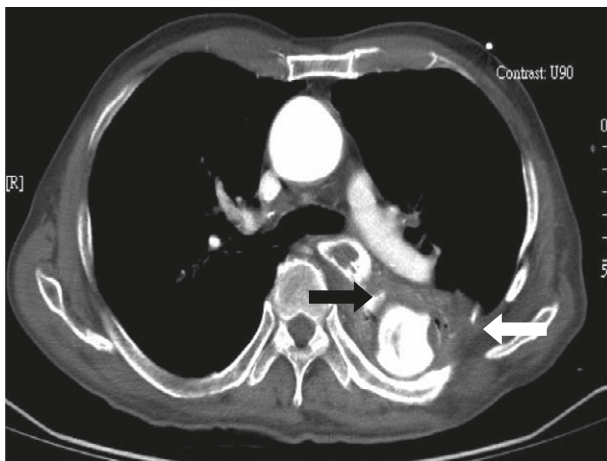


Figure 1. Computed tomography angiography shows contrast extravasation from the descending aorta into the mid-third esophagus (black arrow) and the presence of gas around the aortic prosthesis (white arrow).

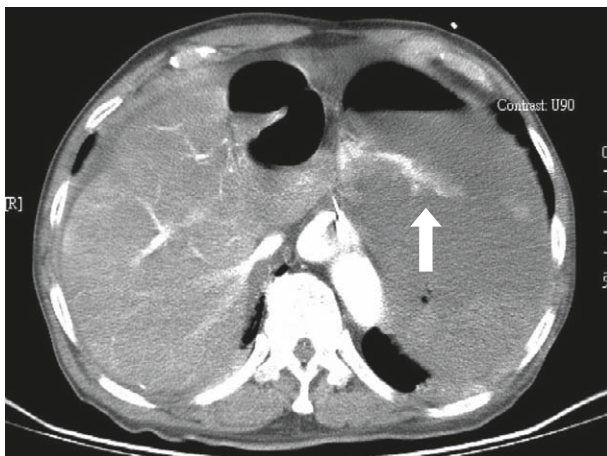


Figure 2. Computed tomography angiography shows contrast medium in the fundus of the stomach (white arrow).

investigation of choice for upper gastrointestinal bleeding, but its definitive diagnosis rate of AEFs is between 20% and 50%.^{10,11} Complicating factors in the endoscopic diagnosis of AEF include an inability to identify a bleeding point as a result of the large volume of blood in the esophageal lumen or a small fistula caused by a foreign body. CTA could show the underlying dissection or aneurysm of the thoracic aorta, periaortic hematoma, esophageal perforation and foreign body, mediastinal abscess or esophageal malignancy. Abnormal adherence of the esophagus to the aorta or aortic aneurysm or the presence of gas in the aneurismal sac or around the aortic prosthesis in the presence of clinical features is virtually diagnostic for AEF.¹² Contrast extravasation from the descending aorta into the mid-third esophagus should be highly suggestive of AEF, as in our case (Figure 1).

AEF should be suspected in patients with aortic graft who present with hematemesis, patients who have undergone aortic and esophageal surgery, and patients with a history of foreign body ingestion. Management depends on early recognition, a high index of suspicion, improved critical care and emergency surgical intervention.¹³ Upper gastrointestinal endoscopy remains the diagnostic study of choice, but CTA should be promptly performed on patients who are suspected of having AEF with obscure upper gastrointestinal bleeding. Rapid AEF identification is important for definitive treatment and improved survival.

References

1. Hollander JE, Quick G. Aortoesophageal fistula: a comprehensive review of the literature. *Am J Med* 1991;91:279–87.
2. Lam ECS, Brown JA, Whittaker JS. Oesophageal foreign body causing direct aortic injury. *Can J Gastroenterol* 2003;17:115–7.
3. Cairols MA, Izquierdo LM, Barjau E, Iborra E, Romera A. Primary aorto-oesophageal fistula due to oesophageal carcinoma: report of a successfully managed case. *Int Angiol* 2000;19:290–3.
4. Robbs JV, Bhoola KD. Aorto-oesophageal fistula complicating tuberculous aortitis (case report). *S Afr Med J* 1976;50:702–4.
5. Hance K, Hsu J, Eskew T, Hermreck A. Secondary aorto-oesophageal fistula after endoluminal exclusion because of thoracic aortic transection. *J Vasc Surg* 2003;37:886–8.
6. Dubrueil JM. Observation sur la perforation de l, esophage et de l, aorte thoracique par une potion d, os avale: avec des reflexions. *J Univ Sci Med* 1818;9:357–65.
7. Chiari H. Ueber Fremdkorperverletzung des oesophagus mit aortenperforation. *Berlin Klin Wochenschr* 1914;51:7–9.
8. Dossa CD, Pipinos II, Shepard AD, Ernst CB. Primary aortoenteric fistula: part II. *Ann Vasc Surg* 1994;8:207–11.
9. Baron RL, Kochler RE, Gutierrez FR, Forrest JV, Weyman PJ. Clinical and radiographic manifestations of aortoesophageal fistula. *Radiology* 1981;141:599–605.
10. Montgomery RS, Wilson SE. The surgical management of aortoenteric fistulas. *Surg Clin North Am* 1996;76:1147–57.
11. Champion MC, Sullivan SN, Coles JC, Goldbach M, Watson WC. Aortoenteric fistula: incidence, presentation, recognition and management. *Ann Surg* 1982;195:314–7.
12. Tierney LM Jr, Wall SD, Jacobs RA. Aortoesophageal fistula after perigraft abscess with characteristic CT findings. *J Clin Gastroenterol* 1984;6:535–7.
13. Chen IM, Chang HH, Hsu CP, Lai ST, Shih CC. Ten-year experience with surgical repair of mycotic aortic aneurysms. *J Chin Med Assoc* 2005;68:265–71.